CHILDREN IN A TUBERCULOSIS COLONY

A SURVEY OF THE PAPWORTH CHILDREN

(Based on the clinical records of Dr. L. B. Stott)

BY

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The demand for statistics about the degree of infection among the Papworth children has been insistent. Sir Pendrill Varrier-Jones himself was always most anxious to supply the evidence required, but both he and Dr. L. B. Stott, who had been in charge of the children’s clinic since the foundation of the settlement, were reluctant to produce any statement before a reasonable period of observation had elapsed. The present writer has been in contact with Dr. Stott since 1926, and has been privileged to follow the elaborate and painstaking method of observation which he introduced with the object of presenting one day the epidemiological picture of the village population. In 1936 Sir Pendrill Varrier-Jones decided to mark the twentieth birthday of the Papworth Village Settlement by preparing the first statistical survey, and the work has been completed and presented in a monograph which is shortly to be published. Here some results of the observations, which will be found in detail in the monograph, are submitted.

The material

Papworth Village Settlement is a community which has grown from small beginnings: from eight cottages in 1918 to 142 cottages in 1938; and the population in 1938 (twenty years after the settlement had been transferred from Bourn to Papworth) consisted of 117 ex-patients’ families. During this period 199 ex-patients’ families occupied cottages in the village: of these, 137 were families with children while 62 were childless. Of the 137 families, 77 are still resident, 33 have left the village following the death of the head of the family, and 27 have left Papworth for various other reasons—seven of them because the patient had completely recovered his health.

The period of residence was less than 5 years in 54 families: from 5 to 10 years in 43 families: 10 to 15 years in 26 families: and 15 to 20 years in 14 families.

In 57 families the head of the family was a persistently sputum positive case: in 63 families the head of the family had become ‘sputum-converted,’ and in 15 families surgical tuberculosis was present. The heads of two families suffered from a non-tuberculous but disabling lung disease.

The number of children in these 137 families were 1-2 in 61.5 per cent.; 3-4 in 25.5 per cent.; and 5-8 in 20 per cent.

Children born in the village

There were 108 children born in the village: twenty-four in families of the healthy staff, and eighty-four in families of ex-patients. Eighteen babies were born in families of sputum negative cases; sixteen in families of surgical cases; twenty-five in families of persistently sputum positive cases (in sixteen of which the father subsequently succumbed to the disease); and twenty-five in families of sputum converted cases. Although the number of cases in each division is relatively small, it is thought advisable to deal with each group separately, as the degree of exposure is naturally different in each group.

Mortality. With regard to the mortality rate we can be brief. Of the 108 children, none have died from tuberculosis. Four of them were stillborn or died a few hours after birth; one died from diphtheria at the age of two, having left the settlement; and another is stated to have died from an attack of laryngospasm after leaving the settlement. Such a low mortality is unique compared with the general child mortality in other groups of the population in town or country; and even compared with other village populations of similar size the child mortality of Papworth must be considered very low.

Case incidence of active lesions. There was no case of active tuberculosis observed among the children independent of the conditions of exposure. This includes extra-pulmonary as well as pulmonary lesions. There was, in fact, no clinical or radiological evidence of active pulmonary or non-pulmonary tuberculosis.

Case incidence of latent lesions. It was expected that in these children some evidence of a previous primary infection would be discovered, and the findings can be tabulated as follows:—

55 (or 51 per cent.) had no clinical or radiological findings.
46 (or 42.5 per cent.) had only radiological evidence of calcified foci (not exceeding those observed in a mixed child population).
5 (or 4.6 per cent.) had radiological evidence of an abortive primary lung lesion.
2 (or 1.8 per cent.) showed evidence of transient perifocal reactions.

The two cases of transient perifocal reaction were
observed in children of sputum positive patients. Of the five cases of residual primary infiltration, three were observed in children born in the families of healthy employees, one in the family of a sputum positive case, and one in the family of a surgical case.

Discussion

It is most gratifying to know that none of the children born in the families of healthy employees or of sputum-negative cases have contracted tuberculosis in an active form when living in village surroundings, and particularly encouraging to find that Sir Pendrill Varrier-Jones’s statement that ‘no child born in Papworth of tuberculous stock has contracted tuberculosis’ has been confirmed. There are reservations to be made. The number of cases in each subdivision made according to exposure is too small actually to calculate percentages; and secondly, in most cases the period of observation covers only a small part of the lifetime of an individual in which manifestations of the disease can be expected. Observations cover the first three years of life, when the acute manifestations are to be seen in children who have been exposed to intrafamilial infection. The question then arises how our observations at Papworth compare with the results of other contact studies. This comparison is made difficult by the fact that there is no actual standard figure, and the figures given for the incidence of active cases and mortality in infants brought up in an environment of open tuberculosis vary considerably.

In 1926 Calmette calculated that the tuberculosis mortality of babies born of tuberculous mothers in their first year of life was 24 per cent. in the provinces, and 32-6 per cent. in Paris. This figure was greatly disputed, and the Paris figures were rectified in 1928 by Hazeman to be 8 per cent. for children of tuberculous mothers in the ages of one month to four years. Other investigators found much lower figures. Jorgensen and Backer (1929) in Oslo found a mortality rate of 7-1 per cent. (196 cases): 6 per cent. for tuberculosis in the first year of life. Deutsch-Lederer (1929) reported a 6-6 per cent. tuberculosis mortality rate; and Braeuning and Neumann (1929) reported 7-5 per cent. (246 cases). In a Lancashire contact study Lissant Cox (1929) found a tuberculosis mortality rate of 3-2 per cent. (from all causes 11-5 per cent.), referring to an analysis of the histories of 1486 children in Lancashire under five years of age, living in 1063 homes, in each of which were one or more adults suffering from open tuberculosis. Dow and Lloyd (1930) found a mortality rate of 1-8 per cent. in 159 contacts 0-1 year old. Turner (1930) found a mortality rate of 3-9 per cent. in 76 contacts aged 1-2 years, from families with a sputum positive fatal case. Frantz of Magdeburg (1930) found much higher figures, reporting 16-9 per cent. in 119 cases.

Ostenfeld and Kjer-Petersen (1931) found a 6-1 per cent. tuberculosis mortality rate in children exposed to infection in infancy and followed up to their third year of life. In 103 children exposed to an open case in the first and second year of life, Scherman (Munich 1931) found 12 per cent. mortality from tuberculosis.

Dieppez (1929) found that out of 888 children born between 1926 and 1930 in households in Cologne where one member suffered from open tuberculosis, 7-2 per cent. died from tuberculosis in the first year of life. The death rate from tuberculosis was 8 per cent. in 382 contact infants reported by Grass of Bremen (1934). Kandziora (1934) found a tuberculosis death rate of 9 per cent. in 65 babies born in tuberculous families.

Seiffert (1935) derived from a questionnaire sent to all the tuberculosis dispensaries in Germany that the tuberculosis mortality of contacts in the first year of life was 11-6 per cent.

Duthoit and Dubois (1936) found a tuberculosis mortality rate of 17-4 per cent. in contact infants living with a sputum positive case for one to twelve months in the first six years of life. Wirtz (1936) recorded 7-7 per cent. deaths from tuberculosis in 168 infants born in the years 1926–1933 in families where the father or mother was a sputum positive case. The mortality rate of the tuberculin positive babies was 11-7 per cent.

Heynsius van den Berg (1936) in Amsterdam found the tuberculosis mortality in children exposed during the first three months of life and followed up for the first two years varying from 2-3 per cent. in good hygienic conditions and slight exposure, to 69-3 per cent. in bad hygienic conditions and massive exposure.

From these figures, collected from all over the world, it can be seen that the tuberculosis mortality of infants born in a tuberculous environment varies to a certain extent, and these variations might be explained by the conditions of exposure.

This is borne out, not only by Heynsius van den Berg’s careful investigations, but Leon Bernard (1928), the French phthisiologist, also insisted upon the fact, and he used to refer in this connexion to the following observation.

In the maternity wards of a hospital for tuberculous mothers the death rate of babies dropped from 38 per cent. in 1921–1923, to 13-2 per cent. in 1923–1925, and to 7-2 per cent. in 1925–1926. This improvement was due to the fact that in the second period a separation of mother and child was assured by keeping them in different rooms, but in contact with their mothers; while in the last period the babies were removed from the building altogether.

Whatever the interpretation might be, the fact remains that the expected tuberculosis mortality of children born in families with an open case varies over a wide range. Most of the figures lie between 2 per cent. and 10 per cent.

It is important to note that the babies born in Papworth do not form a ‘collective’: they are not an aggregate of cases exposed under identical conditions. The exposure varies in the different families, and the number of babies born in the homes of open cases is too small for comparison of statistical calculations. But still there remains the fact that in the scale of investigations Papworth takes the lowest place. This cannot be merely accidental, as at the same time there is a complete absence of other manifestations of activity, such as primary in-
filtrations or glandular enlargements. The incidence of active cases of tuberculosis is much higher than the mortality, though reliable figures about morbidity are not easily obtainable.

One question cannot be answered satisfactorily, that of the infection rate. Dr. Stott had subjected the children to the Moro test as a routine method, and his results were reported by Bardswell in 1933 at the Annual Conference of the Tuberculosis Association at Cardiff. Stott found 63 per cent. positive reactors in the second year of life, or 56.8 per cent. in the age group one to five (referring to 91 cases aged one to five). This figure is on the low side compared with other statistics. Heynsius van den Berg assessed the infection rate at 71.5 per cent. in contact infants in the first year. The infection rate in children exposed in the first year of life and observed during the first two years, varied from 31.2 per cent. in good hygienic conditions and slight exposure, to 100 per cent. in bad hygienic conditions and massive exposure. Seiffert, in his mass survey, found 71.6 per cent. to 79.3 per cent. positive reactors in child contacts under five years old, and the figures of other investigators are on a similar scale.

It might, therefore, be said that the infection rate and the extent of the infection among the babies born in Papworth corresponds to that observed in tuberculous families where almost ideal home conditions prevail.

Children admitted to the village

After this discussion of the children born in Papworth, there remains the bulk of the childhood population which has been admitted to the village with their families at various ages, and who were exposed to intrafamilial contact infection before admission. These are families which have been transferred from their former environment to Papworth after the damage has been done, and there are, therefore, two aspects to be discussed: (a) the spread of infection among these families as a result of exposure before admission, and (b) the effects of the new surroundings upon the development of primary infection.

The standard method for examining the spread of infection in families where one of the parents is the source of infection is the family chart. There are several ways of working out such a chart which was first applied by the late Sir Arthur Newsholme. A very well-known example is the chart worked out in Opie's contact studies at the Phipps' Institute at Philadelphia. The principle of all these charts is the chronological follow-up of all the members of the family in relation to their exposure, and the recording of all the clinical and radiological observations made during the actual period of exposure, and the subsequent follow-up of each individual member.

The type of family chart used in the Papworth survey can be seen from a few examples reproduced when discussing the extent of family epidemics that victimized a number of families before admission to the settlement. Each member of the family is designated by a capital letter (A=father; B=mother; C=first child, and so on). The years are marked horizontally, and each member of the family appears vertically. Negative sputum is indicated by two parallel lines; active disease with positive sputum is indicated by blacking out the space between these lines. If no sputum records are in existence, the upper line is broken; if there is active surgical disease, both lines are broken. The circles on the lines of the dependents indicate the dates when x-ray films were taken, and the number inside the circle, together with the shading refers to the system of case-type definitions which are explained at length in the monograph. Each x-ray film has been reproduced on to a small Leica strip, and from these strips copies have been made on the diagrams. The x-ray films are identified as follows: 1A 28 = the first x-ray taken of the father in 1928; 2A 30 = the second x-ray of the father taken in 1930, and so on. The time of entrance to the village is indicated by the vertical line.

Family charts were made for 239 families who resided in the village, including the families of the healthy staff.

Family epidemics. In a number of families there was a family epidemic in evidence before admission to the village. Family charts 37, 42, 64 and 122 are extreme examples (fig. 1-4). Conjugal infection had occurred in thirteen families. There were ten cases of open tuberculosis, and four cases of closed pulmonary tuberculosis observed in the wives of patients, suggestive of conjugal infection. This would amount roughly to 14 per cent. total, or 10 per cent. open cases among conjugal contacts, if we consider only conjugal contacts who were exposed to an open case.

This indicates a rather higher rate of conjugal infection compared with the average figures reported in the literature. These figures vary considerably. Arnould (1933) in a mass survey on 5369 cases reported 8.4 per cent.; Opie (1932) in a survey on 535 cases reported 12.5 per cent. The figures observed in Papworth, taken in conjunction with the impressive evidence of family epidemics in admitted families, are an indication that the families which applied for and received admission to Papworth were representative of the average contact population.

This is further shown by the mortality and incidence of active and latent lesions in the children of these families. There are 151 children in this group: 76 were under five years old on admission; 67 were of school-age (6-14); and eight were over fourteen years of age. The highest incidence of active childhood lesions was observed in 112 children below the age of ten years: three cases of active childhood tuberculosis and 31 cases of x-ray findings suggestive of residual primary infiltration. Among the 39 children over ten years of age, there was only one case of active childhood tuberculosis: two cases of residual primary infiltration; but nine cases of adult phthisis.

These are indeed record figures. None of the active childhood type of lesions developed during the stay at Papworth; the adult type lesions were in nine cases present on admission, and four other cases...
FIG. 1.

FIG. 2.
developed recently while under observation in Papworth.

In order to give a more accurate picture of the course of contact infection in these children the example of Opie and McPhedran has been followed, and the children grouped according to their age on exposure, and each group followed up separately, the interval between exposure and the first clinical signs of active lesions being recorded.

Among the 116 children who were 0–4 years at the beginning of contact:

28 had no clinical or radiological evidence of lesions;
7 had Ghon foci;
47 showed the usual type of calcification;
26 had some radiological evidence of a residual primary infiltration;
2 showed perifocal reactions;
4 had active childhood lesions;
2 showed symptoms of early adult phthisis.

The incidence of latent childhood lesions characteristic of a preceding primary infection is 20-6 per cent.; of active childhood tuberculosis 3-4 per cent.; and of early adult lesions 1-8 per cent. Those free from clinical or radiological symptoms are 24 per cent.

The next group of children, whose ages were 5–9 years at the beginning of contact, is comparatively small, and includes only twenty-three children.

There is, of course, no statistical calculation possible about the incidence of lesions, but it is evident that in this group the picture so characteristic of a preceding primary infiltration is practically absent. There is one case of perifocal reaction; no case of childhood tuberculosis, but there are four cases out of 23, or 17-4 per cent., which show signs of more or less progressive adult phthisis.

The next group with the contact age of 10–14 years is too small from which to draw any conclusions, and the same applies to the group 15–25 years old at the time of exposure. It must be noted, however, that the three cases known as adult contacts were also suffering from adult phthisis.

**Discussion**

It is not in any way surprising that the infection rate and case incidence in the children exposed before admission to Papworth shows the alarming picture so familiar to the student of contact epidemiology. The figure for active lesions is deceptive, as no cases were recorded here who had died before the family came to Papworth or who did not come with the family for any reason. It is not of so much interest to know how many children had active tuberculosis when they arrived at Papworth as how many developed lesions in later years while under observation in the settlement. In other words, was the rate of exacerbation that to be expected in contact children, or was there any variation from the expected figures?

It is now generally recognized that contact infection in childhood often leads to the reaction which is known from the description of Kuss and Ghon as primary, or initial childhood tuberculosis. The majority of the contact children showed manifestations of such primary infection. The great majority had only latent lesions, but nevertheless might develop the adult type of phthisis after an interval of varying length. If contact infection takes place in later childhood, or in young adult life, the signs of primary infection are usually less obvious; while the development of adult type phthisis follows after a much shorter interval. The importance of this time interval has been shown by Jessel (1930), Macpherson (1936), and especially by Opie and McPhedran (1932) in their extensive contact studies on family contacts. It is there shown that in contacts exposed between 0–9 years of age the mean frequency of tuberculous lesions indicative of a clinically manifest tuberculosis is 3-95 per cent. in the three years following exposure; and 13-45 per cent. in cases 10–14 years after. Thus infection in childhood produces a rise in the incidence of active childhood lesions in the years following exposure; then comes an interval when active lesions are rare; and this is followed by a peak in the incidence of adult type lesions. If the infection takes place in young adult life (fifteen or higher) there is a rising incidence of active lesions from 2-69 per cent. in the year following exposure to 10-59 per cent. 4–5 years after exposure, and to 19-05 per cent. 10–15 years after exposure.

If this picture is accepted as showing the trend of the development of contact infection in childhood contacts, it is reasonable to expect in the children admitted to Papworth the appearance of adult type lesions. In fact, such exacerbations were observed in only two cases in 0–4 years contacts, both quite recently, and one exacerbating while serving with the Forces. Although 30 of the 116 children who were 0–4 years at the beginning of contact have now reached puberty, only two cases of adult phthisis were discovered in addition to the two which were present on admission. This figure is definitely below the expected one, but it is not inferred that residence in the village settlement has been responsible for this satisfactory result. In the two other cases where active and fatal pulmonary tuberculosis developed in recent years, one was a four to nine years' contact, the sister of one of the cases mentioned before. The other was a case of congenital heart disease who suddenly developed fatal adult type phthisis.

It can thus be seen that the picture observed in Papworth fits in quite well with the general picture obtained from other contact studies, with the one difference that since the families took up residence in Papworth the rate of adult type exacerbations was not alarming, and might have been even less if war conditions had not had their repercussions in one way or another on the village life. The point must be stressed that out of the four cases of late exacerbation only one (the case of congenital heart disease) actually occurred in the village, the other three being recorded in one case during military
service, and in the other two cases after the families had left the village. A full description of all the case types observed is given in the monograph.

Case types

It is important not to be too dogmatic about the interpretation of the different stages of contact infection. From the clinical point of view three phases in the development of contact infection must be distinguished: the phase of initial infection, the interval, and the phase of phthisical development. It is not in every case possible to discern all the three phases. There is in the initial lung lesion following exposure, a typical picture in children, but also seen in adults: there are interval symptoms of a more or less definite nature; and finally there are the signs of the development of phthisis. In other cases there is no visible initial reaction, and in others again there may be no interval symptoms.

Several theories have been put forward to account for the protracted character of contact infection. There is no doubt that in a great many cases phthisis in adult contacts develops in an apparently healthy lung without any other preliminary signs of symptoms, except the fact of exposure. These cases have always been a problem. It has been shown that in such cases x-ray films have appeared normal up to three to six months before the actual onset of adult phthisis.

There are cases, however, in which continuity of development can be followed on the x-ray film, and in the two following examples the development from the initial lesion to the final phase of adult phthisis could be watched.

Family 70. In this family the father and daughter were both suffering from pulmonary tuberculosis. The son was still a child when his father developed the disease, and of school age when his sister was discovered to be an open case. An x-ray film taken at the age of eight years showed some calcified foci in the left hilum. When he was fourteen years old an oblong calcified lesion was present in the left middle zone, also a group of calcified foci in the right hilum. These were definite indications of an active initial lesion. The x-ray findings were closely followed up, and two years later a soft, partly calcified focus, 1 inch wide, was recorded in the left apex with visible streaks leading to the hilum. There was no sign of any further developments, and he was accepted by the Army at the age of twenty. Two years later he broke down, and was readmitted to Papworth. A cavity was present in the left upper lobe on the site of the apical focus seen in the previous x-ray films.

Family 96. In this family the father developed open tuberculosis and finally succumbed. One daughter, thirteen years of age on admission, was suspected of having an active initial lung lesion. Calcification developed in the course of time, and when she was twenty years old a number of hard foci were seen in the left apical region. In the following years a definite spread developed in this zone, and the case was classed as a case suspect of pre-phthisical symptoms. The family left the village and the girl married and had a child. She was readmitted to Papworth in 1940 with a large cavity in the left upper lobe at the site of the former lesion. She finally died in hospital. One brother developed acute tuberculosis at about the same time; his case falls into another category and will be discussed elsewhere.

It is most probable that this type of evolution is the rule, although not always demonstrable in the clinical or x-ray findings. Anatomical investigations at Papworth confirm the view lately taken by Sweany, that the contact lesion which develops after exposure might produce by its insidious but continuous growth all the symptoms that we observe during the interval. There is apparently no essential but only a gradual difference between the development of a contact lesion in children and adults.

Conclusions

The following table shows the incidence of lesions in the various groups of children admitted to the village, compared with that in the village-born children up to 1938.

It will be seen that the village-born children and the children of the healthy staff show the same low incidence; while the children in families of sputum negative and surgical cases show perhaps a slightly increased incidence. In contrast to this the children

| TABLE COMPARING THE INCIDENCE OF LESIONS IN THE VARIOUS GROUPS OF ADMITTED CHILDREN AND IN THE GROUP OF CHILDREN BORN IN THE VILLAGE (ALL FAMILIES INCLUDED) |
|-----------------|------------------|----------------|-----------------|------------------|
| Type of lesion  | No clinical or radiological evidence | Ghon foci | Calcified foci | Residual of primary infection | Transient perifocal infection | Childhood tuberculosis | Adult phthisis | \( \sum \) |
| Children admitted in families of | Healthy staff | Sputum neg. and surgical cases | Sputum pos. cases | Village-born children (all families) |
| No clinical or radiological evidence | 13 (40·9 per cent.) | 25 (32·5 per cent.) | 37 (24·5 per cent.) | 55 (51 per cent.) |
| Ghon foci | 2 (6·1 per cent.) | 6 (7·8 per cent.) | 9 (5·9 per cent.) | 6 (5·5 per cent.) |
| Calcified foci | 16 (50 per cent.) | 43 (55·8 per cent.) | 64 (42·4 per cent.) | 40 (37 per cent.) |
| Residual of primary infection | 0 | 1 (2·6 per cent.) | 25 (16·6 per cent.) | 3 (4·6 per cent.) |
| Transient perifocal infection | 1 (3 per cent.) | 2 (2·6 per cent.) | 3 (2 per cent.) | 2 (1·8 per cent.) |
| Childhood tuberculosis | 0 | 1 (1·3 per cent.) | 4 (2·7 per cent.) | 0 |
| Adult phthisis | 0 | 0 | 9 (5·9 per cent.) | 0 |
| \( \sum \) | 32 | 77 | 151 | 108 |
of sputum positive cases show, as would be expected, a high degree of infection, indicating that contact infection has produced a kind of contact epidemic with a great variety of lesions, before they came to Papworth.

With regard to prognosis, more time must elapse. It has been shown that in contacts the exacerbation rate in later age groups depends on the activity of infection in childhood. In a contact population, if the active childhood lesions are very numerous high figures for adult phthisis have to be expected. If, as in the case of the village-born children, the number of active initial lesions are extremely few, a rise in adult exacerbations need not be feared so long as the resistance of the individual and the environment are under control.

REFERENCES

Seiffert, E. (1935). Ibid., 73, 162.